# Hemosuccus Pancreaticus

## An Unusual Cause of Upper Gastrointestinal Bleeding

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Two cases of hemosuccus pancreaticus as the source of upper gastrointestinal bleeding of unknown cause are presented. The literature on this rare cause of gastrointestinal blood loss is reviewed. Also, the influences of time-course and clinical appearance on the diagnosis of this problem are described.

INTERMITTENT GASTROINTESTINAL BLEEDING of unknown cause is often a diagnostic problem to even the most astute clinician. Our recent experience with two patients who had acute intermittent gastrointestinal blood loss caused by hemosuccus pancreaticus enlightened us about this most unusual clinical syndrome. "Hemosuccus pancreaticus" is the term coined to describe the syndrome of gastrointestinal bleeding into the pancreatic duct manifested by blood loss through the ampulla of Vater.[1] In previous reports on gastrointestinal blood loss of pancreatic cause, the point of entrance of the blood into the gastrointestinal tract has not been well-demonstrated. The origin was usually assumed to be in an erosion of the bowel wall by a pathologic pancreatic process and not, as was the case in our patients, in the pancreatic duct itself. We present reports of these two patients, along with some pertinent clinical points they served to illustrate.

## Case Reports

**Case 1.** A 62-year-old woman was referred to the Mayo Clinic in September 1983 for evaluation of recurrent upper gastrointestinal bleeding of 19 months' duration.

Her problems began in February 1982. She was admitted to her local hospital with symptoms of pallor, weakness, and melanotic stools. Her hemoglobin value was 2.5 g/dl at admission, and she received a transfusion of 5 units of packed red blood cells. Evaluation at that time included a proctoscopic exam and a barium upper gastrointestinal examination, the results of which were negative. A barium enema examination revealed a 1-cm polyp at the splenic flexure and two small polyps in the descending colon. These were removed during colonoscopy, which did not detect any other abnormalities.

Dyspnea recurred in August 1982, and at that time her hemoglobin value was 6.5 g/dl. She was not aware of acute bleeding. A bone marrow examination revealed mild hypercellularity and decreased iron stores consistent with chronic blood loss. She was treated with transfusions and oral iron therapy.

In May 1983, she had lower abdominal pain, diarrhea, and melanotic stools. Her hemoglobin value was 4.6 g/dl. Results of repeat upper gastrointestinal and colon examinations were negative, as were those of an upper gastrointestinal endoscopic examination. In June, exploratory celiotomy along with intraoperative colonoscopy was performed. The endoscopy revealed blood in the cecum; the patient underwent vagotomy and pyloroplasty along with a right hemicolectomy for possible thickening at the appendiceal stump. Pathologic examination of the specimen revealed a 0.3-cm polyp in the colon and a few diverticula. Two months later, dyspnea and fatigue returned, and she was treated with oral administration of iron after a hemoglobin value of 5.9 g/dl was found.

In early September, the hemoglobin level was still low (5.7 g/dl), and a test for occult blood in the stool was positive. An upper gastrointestinal endoscopy found a superficial gastric erosion that was not bleeding and was benign on biopsy. Attempted colonoscopy was limited by sigmoid adhesions. As the month progressed, her dyspnea worsened and her hemoglobin value decreased to 5.1 g/dl but she denied any symptoms of gastrointestinal blood loss. At this point, she was referred to our center for evaluation.

At admission, the patient had a hemoglobin level of 8.3 g/dl and her platelet count was 218,000. Folate and vitamin $B_{12}$ levels were normal; results of serum iron studies and a peripheral blood smear were compatible with chronic blood loss. Findings on proctoscopic exam and a colonoscopy to the level of the ileoascending colostomy were negative. During esophagogastroduodenoscopy, performed to a level 15 cm distal to the ligament of Treitz, neither bleeding nor potential lesions were seen. Upper gastrointestinal and small bowel examinations were remarkable only for a midesophageal diverticulum. At a follow-up determination, the hemoglobin level had decreased to 7.9 g/dl, in spite of no evidence of active blood loss. Hematologic evaluation at this time revealed evidence for chronic blood loss only.

A visceral angiogram revealed a small pseudoaneurysm of the pancreatic magna branch of the splenic artery (Fig 1) and three small vascular ectatic foci in the rectum and distal portion of the sigmoid. Flexible sigmoidoscopy, repeated to examine the rectal vascular lesions,
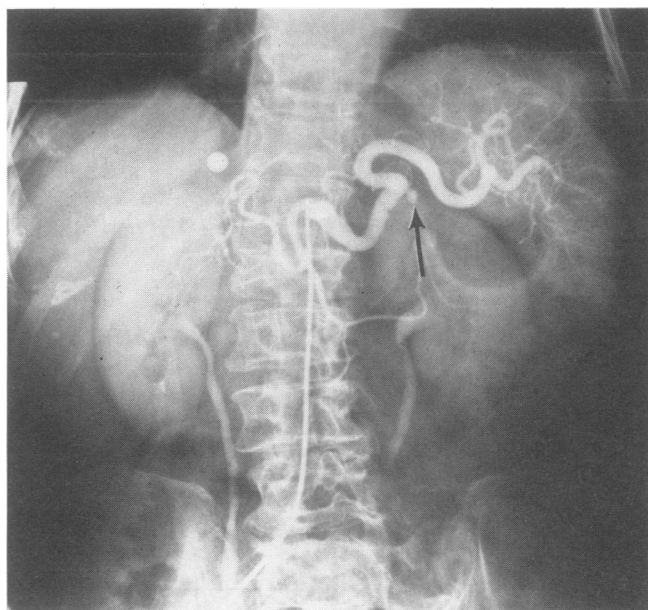
FIG. 1 (Case 1). Visceral angiography. Pseudoaneurysm is indicated by arrow.

was again unrevealing. A computed tomographic scan of the abdomen revealed only an atrophic-appearing pancreas.

That evening (September 28) she passed a large maroon stool that contained blood clots. An emergency scan with technetium-tagged red cells was negative for a bleeding site. Endoscopic retrograde cholangio-pancreatography revealed an 8-mm focal filling defect in the midpancreatic duct, with dilatation of the proximal duct (Fig. 2). This finding was considered compatible with a stone, a clot, or an extrinsic defect from the small pseudoaneurysm.

While exploration was being considered, a bowel preparation protocol was begun, and the patient passed maroon stools when given enemas. Colonoscopy was performed at once, and fresh blood was seen to be coming from above the previous anastomosis. Upper gastrointestinal endoscopy was then done, and blood was seen to be issuing forth from
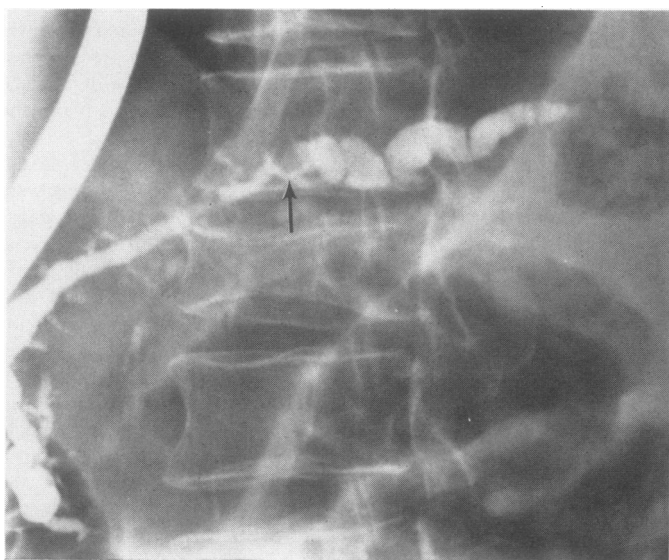


FIG. 2 (Case 1). Endoscopic retrograde cholangiopancreatography. Discrete filling defect (arrow) represents clot within pancreatic duct.

the ampulla of Vater. With this information, operation was then undertaken and a distal pancreatectomy and splenectomy were performed. Extensive scarring and changes of chronic pancreatitis were found at surgery. On pathologic examination of the specimen, a small pseudoaneurysm of the splenic artery was found to be communicating with the duct of Wirsung (Fig. 3).

Case 2. A 64-year-old man was transferred to our institution for evaluation of gastrointestinal bleeding of obscure origin. Five years earlier, he had had his first episode of crescendo epigastric pain. The pain was confined to the epigastrium and was described as "a balloon expanding" in that region. The pain would become very intense within about 45 minutes and was associated with mild nausea, but no vomiting. The pain would then spontaneously decrease in intensity for about 1 hour until totally gone. Several hours after the pain, when the patient had become entirely asymptomatic, melena and hematochezia would develop, and they would persist for 2 to 4 days.

He experienced one such episode each year, until 2 months before coming to the Mayo Clinic; in that period, he had four or five episodes identical to those previously described. He had been hospitalized on several occasions and had undergone multiple intestinal contrast studies and endoscopic procedures to determine the site of his bleeding. The only positive finding had been esophagitis secondary to hiatal hernia with esophagogastric reflux.

After admission to our hospital, the patient had the same crescendo-decrescendo epigastric pain, and several hours later, hematochezia occurred. During this episode, the patient's serum amylase concentration increased to 292 U/liter (normal, 3–23 U/liter), and the serum lipase value increased to 201 U/liter (normal, 4–24 U/liter). His white blood cell count was 5900/$\mu$l, and his hemoglobin value decreased from 12.5 g/dl to 9.8 g/dl. An abdominal radiograph was remarkable for a 4-cm calcified, rounded density in the left upper quadrant (Fig. 4). The results of physical examination were negative.

Because of our recent experience with the patient in Case 1, a working diagnosis of hemosuccus pancreaticus was entertained. A visceral arteriogram revealed a 4-cm splenic artery aneurysm near the splenic hilus, but did not show extravasation into the pancreatic duct (Fig. 5). An endoscopic retrograde pancreatogram revealed incomplete filling of the distal pancreatic duct and no communication with the splenic artery aneurysm (Fig. 6).

Surgical exploration was undertaken, and a distal pancreatectomy and splenectomy were performed. The specimen revealed a large true splenic artery aneurysm communicating with the pancreatic duct, which ended at the medial aspect of the aneurysm (Fig. 7).

## Discussion

Hemosuccus pancreaticus is a syndrome of blood loss through the duct of Wirsung, and is usually accompanied by acute relapsing or chronic pancreatitis. The disease is unusual, only 18 cases having been gleaned from the literature in our review. Although bleeding into a pancreatic pseudocyst is somewhat more common, the term "hemosuccus pancreaticus," defined by Sandblom[1] in 1970, is properly applied only to blood loss that occurs into the gastrointestinal tract through the ampulla of Vater. In his report of three cases, Sandblom coined the term to describe the similarity of the disorder to the clinical syndrome of hematobilia.

The first case of hemosuccus pancreaticus was described in 1931 in a report by Lower and Farrell,[2] of a 16-year-old boy with several years of intermittent melena and abdominal pain as a sequela of an episode of whooping cough. At surgery, a splenic artery aneurysm
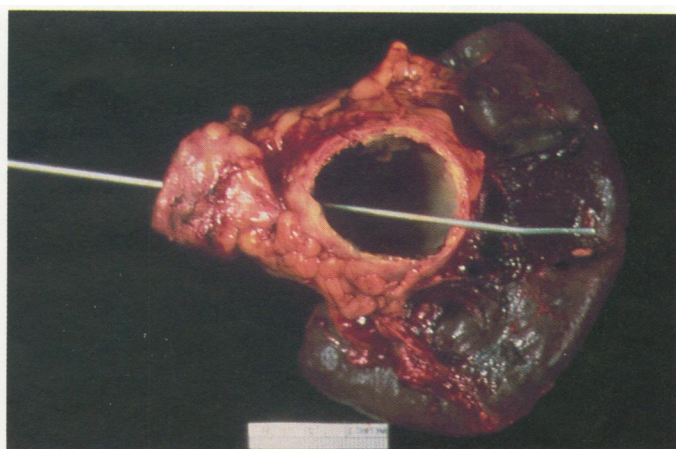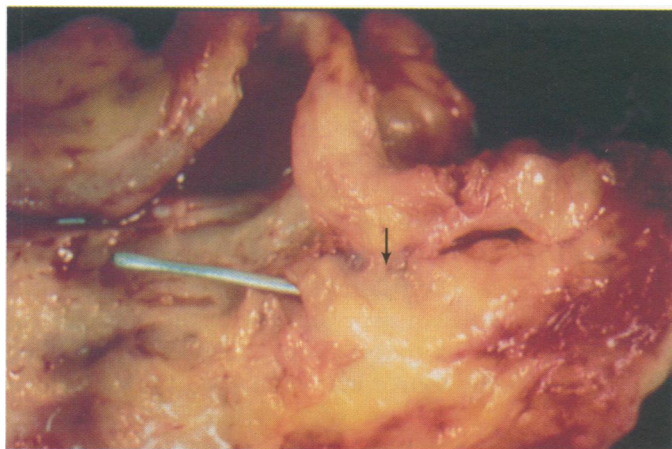
FIG. 3 (Case 1), *left*. Probe lies within pancreatic duct. Arrow marks communication of pseudoaneurysm with pancreatic duct. FIG. 7 (Case 2), *right*. Surgical specimen. Probe lies within pancreatic duct.

communicating with the pancreatic duct was found. Distal pancreatectomy and splenectomy were curative.

It was not, however, until the advent of angiography that pancreatic communication with the surrounding vasculature was appreciated as a significant cause of gastrointestinal bleeding. Three cases were reported in the 1960s and, in 1970 Sandblom[1] published the previously mentioned paper. In this paper, Sandblom reported two confirmed cases of the disease, which he termed "hemosuccus pancreaticus." The first patient was a 73-year-old man with a history of postcholecystectomy pancreatitis who was seen because of abdominal pain and anemia. Continued hematemesis despite negative results of radiologic examination of the gastrointestinal tract led to exploration; the findings were a pulsatile mass displacing the upper part of the duodenum and some diffuse bleeding at the site of a gastroduodenostomy done 3 years earlier, after resection of a gastric carcinoma. The patient underwent postoperative angiography, which demonstrated an aneurysm of the common hepatic artery. Reoperation could not reach the site of bleeding because of inflammation, and the patient died of continued bleeding. At postmortem examination, the hepatic artery aneurysm was found to connect with the duct of Wirsung.
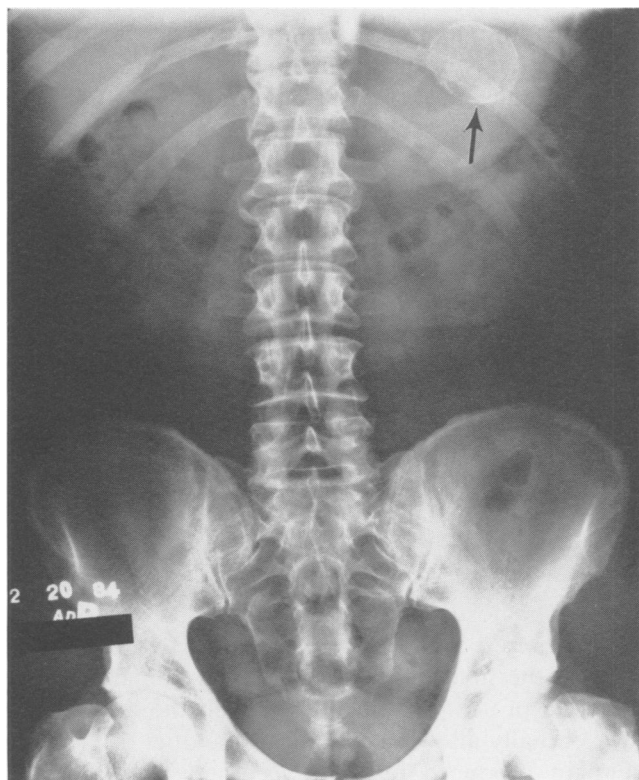


FIG. 4 (Case 2). Abdominal radiograph reveals calcified spherical mass (*arrow*) in left upper quadrant.
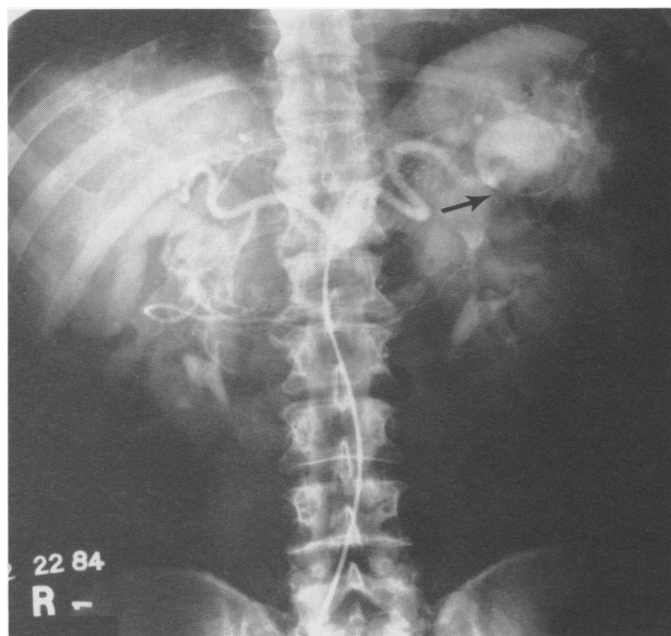


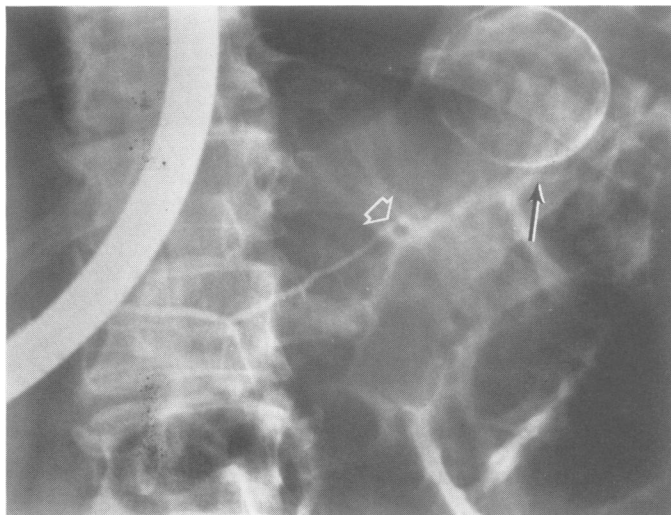FIG. 5 (Case 2). Laminated clot (*arrow*) within splenic artery aneurysm.

FIG. 6 (Case 2). Endoscopic retrograde cholangiopancreatography. Cutoff of pancreatic duct (*open arrow*) is separate from splenic artery aneurysm (*dark arrow*).

Sandblom's second patient was a 51-year-old man with a history of alcohol abuse and a splenic artery aneurysm that was atherosclerotic. The aneurysm was found on angiograms after the man had undergone a blind gastric resection for recurrent gastrointestinal bleeding at a different hospital before being referred to Sandblom. Distal pancreatectomy and splenectomy were performed, and the patient's bleeding resolved. Sandblom also reported a case of suspected, but not proved, bleeding from three small splenic artery aneurysms.

The most recent report, by Cahow et al.[3] in 1983, presented three cases of hemosuccus pancreaticus diagnosed before surgery by angiography. This report is noteworthy not only for the youth of the patients (they were 29, 35, and 47 years of age) but also for the fact that in two patients the pseudoaneurysm was located in the common hepatic artery. These are the first hepatic artery pseudoaneurysms reported as a cause of hemosuccus pancreaticus since Sandblom's report.

In the area of correct preoperative diagnosis, our report matches that of Cahow et al. in being somewhat unusual. The diagnosis of hemosuccus pancreaticus was made before surgery in only two other cases of which we are aware. In 1978, Bivins et al.[4] reported on a case of bleeding from a splenic artery pseudoaneurysm detected at preoperative angiography. Of special note is a report from Brintnall et al.,[5] in 1974, of a patient in whom blood was seen issuing from the ampulla of Vater at endoscopy, the same sign seen in our first case. This is the only other occurrence of endoscopic visualization of this bleeding we have discovered.

The results of endoscopic retrograde pancreatography in the diagnosis of this entity are equivocal. In our first patient, the procedure helped demonstrate a filling defect representing a clot in the duct in a location corresponding to that of the pseudoaneurysm found on angiography, focusing our attention on what had been thought an incidental finding. In the second patient, however, the apparent cutoff of the duct, which we assume was produced by clot filling its distal portion, was misleading. It appeared that the pancreatic duct was not anatomically related to the calcific density representing the splenic artery aneurysm. The patient's highly suggestive clinical history and our recent experience with the first patient, however, caused us to persevere in believing that the aneurysm was the source of bleeding *via* the pancreatic duct.

Other than in a report by Longmire and Rose[6] of a young girl with hemosuccus pancreaticus caused by a gastric and duodenal duplication that ulcerated and bled through the ampulla of Vater, the cause of this disease has always been identified as an aneurysm or pseudoaneurysm of a visceral artery. In cases without a pseudocyst, this has been the splenic artery or one of its smaller branches, usually the pancreatic magna artery. The single exception is Sandblom's first case, in which a pseudocyst was not mentioned and the aneurysm was in the hepatic artery. When a pseudocyst has been present to cause erosion of a vessel, the hepatic, gastroduodenal, and pancreaticoduodenal arteries have been found to contain pseudoaneurysms.

In understanding the pathogenesis of this problem, one must differentiate between aneurysms and pseudoaneurysms. In only two cases has the cause of hemosuccus pancreaticus been documented as a true aneurysm of the splenic artery, atherosclerotic in nature. Our second patient had such an aneurysm, as did Sandblom's second. Although the character of the vessel wall dilatation has not always been defined in previous case reports, the defect has been described in most as a pseudoaneurysm that seems to originate from enzymatic destruction of the arterial wall during a bout of acute pancreatitis. Walter et al.[7] stated that up to 10% of patients with chronic pancreatitis have such pseudoaneurysms in surrounding visceral arteries.

In contrast to splenic artery aneurysms, pseudoaneurysms occur most often in men, are not etiologically related to pregnancy, and seldom rupture in a single catastrophic event.

In the tendency of pseudoaneurysms to produce a prolonged history of intermittent bleeding, the natural history of hemosuccus pancreaticus can be seen. In most cases, a severe bout of pancreatitis precedes, sometimes by years, the first episode of bleeding. Apparently, the pancreatic proteolytic enzymes breach the arterial wall and eventually allow bleeding into the duct of Wirsung. The pain produced by this bleeding is secondary to rapid ductal distention, which also causes the leak to seal as pressure in the duct rises. This development is

well-demonstrated in our second patient's course of crescendo-decrescendo abdominal pain. The ductal blockade engenders recurrent pancreatitis and produces an elevated serum amylase concentration.[8] As the duct is decompressed through the ampulla of Vater, clinical gastrointestinal blood loss is seen. Afterward, the pancreas recovers and pancreatic enzymes once again lyse the clot that has sealed the leak, and the process repeats itself.

Logical analysis of this pathogenesis leads to the conclusion that, to demonstrate extravasation through the pancreatic duct, one should perform angiography during the patient's attack of pain, because this is when bleeding is actually taking place. If angiography is delayed until blood is seen, only the aneurysmal vessel will be found; this discovery leads to clinical suspicion but not to definitive diagnosis. It must be pointed out, however, that this syndrome can be clinically silent, as in our first patient, who fervently denied abdominal pain before surgery and would only grudgingly admit to it in close postoperative questioning.

A variety of procedures have been reportedly successful in this problem, from transarterial electrocoagulation, embolization, or balloon occlusion to Whipple resection.[3,9] In most cases, the procedure of choice has been distal pancreatectomy with splenectomy. Some authors have advocated proximal and distal ligation of the aneurysmal vessel, with either oversewing of the ductal communication[10] or drainage of a pseudocyst, if present. In most cases, the transarterial approaches have been used as temporizing measures or when surgical approach was not feasible. The case for resective therapy as opposed to splenic preservation by aneurysmal ligation was well-outlined by Cahow et al.[3] In leaving behind diseased gland in close proximity to the previously injured artery, one leaves the patient with an organ prone to recurrent pancreatitis and possible reoccurrence of arterial injury and bleeding.

## Conclusion

We hope to highlight several salient points in the cases we have presented.

In the evaluation of a patient with gastrointestinal bleeding of unknown cause, one should always keep in mind that "unknown" does not mean "unknowable." Too often, these patients either are submitted to blind resection of a portion of their gastrointestinal tract or are left to live with their condition—at the least inconvenient and at the worst incapacitating and life-threatening—facing long-term iron therapy and the hazards of intermittent transfusion. In evaluating the patient with gastrointestinal bleeding, one should always re-

member that the pancreas is a part of the gastrointestinal tract and, like all the rest, is prone to blood loss.

Blood loss from hemosuccus pancreaticus can be accompanied by silent pancreatitis. Despite the usual severity of pancreatic pain, our first patient denied abdominal pain of any significance in association with her bleeding. Only the finding of a small pseudoaneurysm on her angiogram led to clinical suspicion and, eventually, to diagnosis of her disease. That she did have pancreatitis was obvious when she was examined at surgery and was emphasized by the difficulty of resection due to peripancreatic fibrosis.

The nature of this rare condition is such that the time-course of the patient's blood loss must be understood for angiography to produce a definitive diagnosis if a bleeding papilla is not observed. Angiography should be performed during the patient's attack of pain, since this is when blood loss is occurring, to document extravasation. As previously mentioned, most reports of pancreatic blood loss through the gastrointestinal tract do not document the exact site of entry of blood into the bowel. Bleeding observed to be occurring through the ampula of Vater is exceedingly rare. Perhaps many of the patients with gastrointestinal bleeding associated with pancreatitis or pancreatic pseudocysts are losing blood in this manner, and careful use of endoscopy or properly timed angiography will prove hemosuccus pancreaticus to be a more common entity than we now believe.

## References

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